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**Case Report**

# **Strangulated Urethral Prolapse in Oriental Girls: A Rare Case Report and Literature Review**

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Urethral prolapse in girls is an uncommon condition usually presenting with edematous, friable rosette of bright red or cyanotic tissue and mainly involves the distal urethra. It has been recognized as a disease of premenarcheal black girls and post-menopausal white women. The etiology is not completely understood. We report here a rare case of urethral prolapse with gangrenous change in a 7-year-old oriental girl.

**Key word:** urethral prolapse

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## **Introduction**

Urethral prolapse is a rare condition with an estimated incidence of one in 3000.<sup>1</sup> It usually presents with edematous, friable rosette of bright red or cyanotic tissue and mainly involves the distal urethra. The underlying cause of this condition remains uncertain. It has been recognized as a disease of premenarcheal black girls and post-menopausal white women in the literature.<sup>2,3,4</sup> Herein, we present a rare case of urethral prolapse with gangrenous change.

## **Case Report**

A 7-year-old Taiwanese girl was referred

from a gynecologist due to perineal pain with a vulvar mass and blood spotting on her underwear. She was quite healthy until she suffered from cough one month before. Besides, no fever, dysuria, frequency, other unusual vaginal bleeding, trauma, gynecologic disorders, nor urinary tract infection was noted. She denied having inserted any foreign body into her vagina.

On physical examination, the vulva appears normal except for a clearly dark-red, doughnut-shaped mass visible at the introitus with serosanguinous discharge (Fig. 1A). After retraction of the labia majora and minora, the mass was found to surround the urethral opening and obscure the hymen. Medical history including birth history was not contributory. Results of medical laboratory studies were

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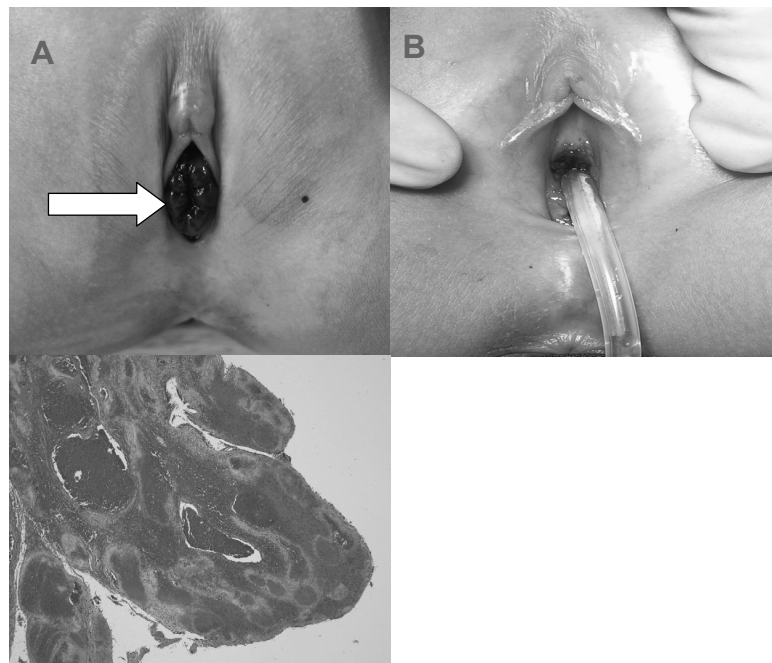
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*Fig. 1 A: The picture of vulvar mass. B: The picture post urethroplasty. C: The pathologic picture showed polypoid tissue partially clothed by urothelium, with severe congestion and hemorrhage.*

also unremarkable. The diagnosis of urethral prolapse was confirmed by gross observation without other specific radiological examination. Due to persistent perineal pain and strangulation of the vulvar mass, excision of the prolapsed urethra and urethroplasty were performed after satisfactory communication with her parents (Fig. 1B). Pathologic analysis showed polypoid tissue partially clothed by urothelium, with severe congestion and hemorrhage (Fig. 1C). Post-operative recovery was uneventful.

## Discussion

The etiology of urethral mucosal prolapse is not well-defined and several mechanisms have been proposed. Capraro et al. described that the changing relation of pelvic organs during the period of accelerated growth of pelvic bones, weakness of urogenital diaphragm in asthenic girls, congenital defect of adhesion between the mucous and submucous membranes, and high intra-abdominal pressure during physical activity may play a role

in provoking urethral prolapse.<sup>5</sup> Anatomic dissection by Lowe et al.<sup>6</sup> demonstrated that urethral prolapse may result from poor adherence between smooth muscle layers of the urethra in associated with episodic increases in intra-abdominal pressure. There is a consensus that increased intra-abdominal pressure is a key factor contributing to the condition. In this case, the young girl had severe, continuous cough persisting for one month. We believed that this was the predisposing factor resulting in the prolapse of urethra in our patient, although chronic cough is an unlikely cause of urethral prolapse in young females.

Literature review showed no report on the incidence of urethral prolapse in oriental girls. Prolapse of urethral mucosa occurs most often in pre-pubertal black girls and in postmenopausal white women.<sup>2</sup> We think that ethnic differences may also play an important role.

The therapy of urethral prolapse includes conservative and surgical treatment. Conservative therapy aims at reducing mucosal edema, improving local hygiene and counteracting

lack of estrogen by using a combination of two or more of the following measures: sitz baths, topical estrogen cream, antibacterial wash/soap and topical antibiotics. Surgical management of urethral prolapse involves excision of the prolapsed mucosa circumferentially. Girls with mild symptoms can be managed conservatively. However, in patients presenting with more severe symptoms or evidence of vascular compromise, surgery is the treatment of choice. The potential complication of surgical treatment is urethral stenosis.<sup>1</sup> In our case, surgical intervention was suitable due to urethral mucosa gangrenous change. Satisfactory post-operative recovery was also noted.

### Conclusions

We report a rare case of a young oriental girl presenting with strangulated urethral prolapse. Since the etiology is still not well-defined, further research is warranted to eluci-

date the underlying mechanism. Moreover, surgical intervention is indicated if conservative treatment fails in achieving satisfactory symptom-control or when tissue gangrene develops.

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